SHORT REPORT

Post-ictal Klüver-Bucy syndrome after temporal lobectomy

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Abstract

In both animals and humans, Klüver-Bucy syndrome is produced by bilateral temporal lobectomy. It is characterised by hypersexuality, visual agnosia, strong oral tendencies, dietary changes, and hypermetamorphosis. Recurrent, postictal Klüver-Bucy syndrome occurred transiently after seizures in a female who had undergone unilateral temporal lobectomy. The pathophysiological mechanism may have been postictal dysfunction of the remaining temporal lobe, producing a transient functional bilateral temporal lobectomy.

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In 1939, Klüver and Bucy described a behavioural syndrome in rhesus monkeys that followed bilateral temporal lobectomy.[1] The syndrome was characterised by prominent and indiscriminate hypersexuality, excessive oral tendencies, loss of normal fear and anger, hypermetamorphosis (excessive attentiveness to visual stimuli), psychic blindness (inability to recognise objects without loss of gross visual discrimination), and dietary changes.

In 1955 Terzian first reported the same syndrome in humans after bilateral temporal lobectomy.[2] Klüver-Bucy syndrome is also associated with a variety of neurological disorders, including herpes encephalitis, Pick's disease, Alzheimer's disease, cerebrovascular accidents, cerebral trauma, and temporal lobe epilepsy. The common feature of all aetiologies was bilateral mesial temporal lobe destruction or dysfunction.

We report an unusual case of recurrent transient Klüver-Bucy syndrome that only occurred during the postictal period in a patient who had occasional seizures after a unilateral temporal lobectomy.

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Case report

In 1984 a 24 year old female sought treatment after generalised seizures followed a week-long fever. A CT scan showed a nonenhancing radioluscent area between the left medial temporal lobe and the left cerebral peduncle that suggested atrophy of the left medial temporal lobe (Fig). Two lumbar punctures were normal. She continued to have frequent seizures and a week later developed status epilepticus despite therapeutic levels of phenytoin, phenobarbitone, and valproic acid. A 24 hour EEG demonstrated that bilateral frontotemporal ictal patterns were occurring every 30 minutes and lasting 70 to 90 seconds. These EEG patterns were associated with the clinically observed seizures. Eighty one per cent of the ictal events originated from the left side.

MRI showed increased signal intensity of the left temporal lobe. Status epilepticus was persistent and refractory to maximal medical therapy. The patient therefore had a left temporal craniotomy 2 weeks after admission. Electrocorticography showed many independent spike discharges from the inferior and middle temporal gyri. A temporal lobectomy removed the anterior 4 cm of the left temporal lobe, the amygdala, and the anterior 1 to 1·5 cm of the hippocampus.

Postoperatively, the patient did well. She experienced no further seizures while receiving phenytoin, phenobarbitone, and carbamazepine in the hospital. An EEG showed spike and wave complexes and sharp waves from the left frontotemporal region. Because of the unusual need for temporal lobectomy to control status epilepticus, this case was reported in the neurosurgical literature in 1986.[3] The patient was then lost to follow up several months after surgery.

Five years later after a generalised seizure, the patient sought treatment in a local emergency room. On examination, she was lethargic and unresponsive but medically stable without further seizure activity. She was left unattended in an examination room. About 30 minutes later, she was found in an adjacent room performing fellatio on an elderly male cardiac patient. The patient was easily led back to her room. She was placid and apparently did not recognise family members who arrived soon after. An hour later she was alert and neurologically intact with no memory of the episode.

When questioned, the patient's family revealed that she still had occasional seizures despite the temporal lobectomy. Typically, the seizures occurred after she had run out of anticonvulsant medication. After the seizures, the family reported that she exhibited abnormal sexual behaviour, which included masturbating in public and soliciting intercourse with family members and neighbours. During these episodes, she did not recognise family members and was subdued and apathetic. She also exhibited bulimia, eating voraciously whatever food was available. These behavioural changes lasted 1 to 2 hours; afterwards, she was amnestic for the entire event. During

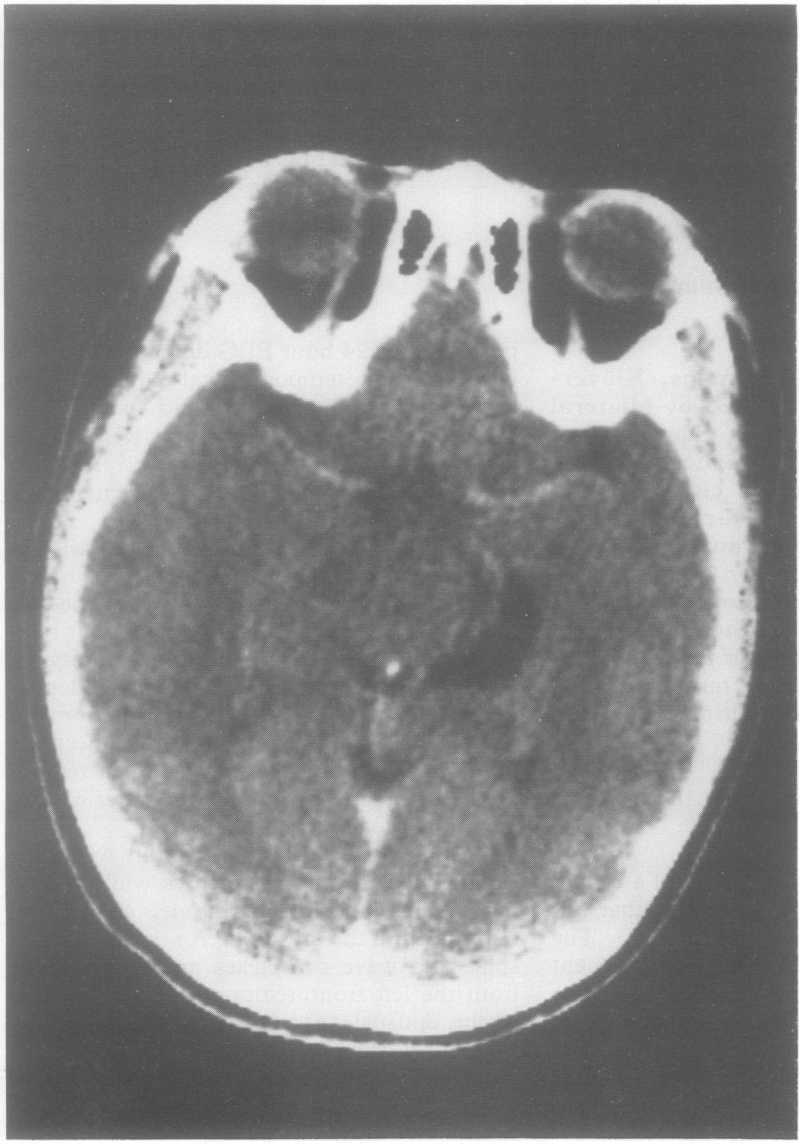

Figure CT scan performed after the patient's initial presentation with status epilepticus demonstrates only a low density area between the left medial temporal lobe and cerebral peduncle suggesting medial atrophy.

her prolonged inter-ictal periods, she behaved normally and exhibited no hypersexual conduct.

Discussion

This patient exhibited most of the behavioural symptoms associated with Klüver-Bucy syndrome, including hypersexuality, loss of normal emotional response, and dietary changes. Her apparent prosopagnosia is consistent with the psychic blindness exhibited by animals with Klüver-Bucy syndrome. This finding has also been reported in other cases of humans with Klüver-Bucy syndrome.[4] The expression of her hypersexuality by performing fellatio on a stranger could also reflect the strong oral tendencies associated with Klüver-Bucy syndrome. Although amnesia is common after seizures, it is also present in most humans with Klüver-Bucy syndrome.[4] Hypermetramorphosis was absent in our case but humans typically show this by exploring the environment manually and by placing objects into their mouth.[4 5]

Most reported cases of Klüver-Bucy syndrome in humans have been associated with a chronic or progressive neuropathological syndrome. The manifestation has rarely been transient.[4 5] Klüver and Bucy noted that in monkeys the behavioural syndrome did not change during two years of follow up.[1] Our patient is unusual because the behavioural symptoms associated with Klüver-Bucy syndrome only appeared transiently during the post-ictal period. Her behaviour was otherwise normal between seizures.

After her temporal lobectomy, this patient's other temporal lobe may have temporarily dysfunctioned after a seizure. The resulting functional bilateral temporal lobectomy presumably caused the Klüver-Bucy syndrome to appear transiently. As her unilateral temporal lobe function reappeared, the symptoms resolved and she behaved normally.

A number of changes in sexual behaviour are associated with temporal lobe epilepsy and its treatment by lobectomy. As many as 71% of the patients with temporal lobe epilepsy exhibit altered sexual behaviour associated with ictal events.[6] Inter-ictal hyposexuality occurs in 80% of the patients; 20% exhibit a variety of paraphilias.[6] When these hyposexual patients have a unilateral temporal lobectomy, their libido frequently increases and becomes excessive in some cases.[7] Conversely, aberrant sexual behaviour as part of the ictal manifestation of temporal lobe seizures is also well documented.[8] Neither of these phenomena seems to account for our patient, whose altered behaviour was clearly confined to the post-ictal period.

Post-ictal hypersexuality after unilateral temporal lobectomy is rare. Blumer noted two isolated cases in his series of 42 patients who had unilateral lobectomy for temporal lobe epilepsy.[7] Only one of the cases is described without mention of other symptoms of Klüver-Bucy syndrome. Consequently, it is not clear if that case represents transient post-ictal Klüver-Bucy syndrome. Cogen *et al* also described a patient who exhibited "partial" Klüver-Bucy syndrome after a unilateral temporal lobectomy.[9] In that case, however, the behavioural syndrome was chronic and unrelated to the post-ictal period.

Post-ictal alterations in sexual behaviour have also been reported after temporal lobe seizures in patients who have not had surgery.[6 7] Perhaps these episodes reflect transient bilateral dysfunction of the temporal lobe after a seizure. Such temporary functional bilateral lobectomy could cause the same syndrome as an anatomical lobectomy and would be similar to the pathophysiological mechanism which may account for our patient's behaviour.

Klüver-Bucy syndrome remains a fascinating syndrome whose exact neuroanatomical basis is unclear. As neurosurgical treatment of seizure disorders increases, the behavioural consequences of mesial temporal lobectomy must be considered. Although most patients

with intractable seizures improve after surgical intervention, they may develop neurobehavioural complications such as the Klüver-Bucy syndrome. A more limited surgical resection, particularly one that spares more of the amygdala, may minimise the chance of this type of complication.

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